Bilateral Tension Pneumothorax and Tension Pneumoperitoneum Secondary to Tracheal Tear in a Patient With Relapsing Polychondritis

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Relapsing polychondritis (RP) is a rare disease that is characterized by recurrent inflammation and destruction of cartilage and connective tissues. RP can have significant airway pathology that may require procedures to maintain airway patency and thus may have serious implications for anesthesiologists. Anesthesiologists must be prepared to deal with the possible complications that may occur during airway manipulation in patients with RP. Here, we present a case of life-threatening bilateral tension pneumothorax and tension pneumoperitoneum that developed after a tracheal tear during Montgomery T-tube insertion in a patient with tracheal stenosis due to RP. Correct diagnosis was delayed due to a misdiagnosis of airway obstruction. As a result, we emphasize that bilateral tension pneumothorax should be considered during refractory cardiac arrest in patients with increased airway pressure. A high index of suspicion and adequate management are mandatory for patients to survive these life-threatening complications. [*J Chin Med Assoc* 2009;72(9):488–491]

Key Words: pneumoperitoneum, pneumothorax, relapsing polychondritis, tracheal stenosis

Introduction

Undiscovered tracheal rupture can cause fatal complications, especially when patients are under positive pressure ventilation. One such fatal complication is bilateral tension pneumothorax that may be difficult to diagnose without a high index of suspicion.[1,2] Tension pneumoperitoneum occurring as a result of tracheal tear is rarely reported in the literature. We present here a case of life-threatening bilateral tension pneumothorax and tension pneumoperitoneum, which developed secondary to a tracheal tear during Montgomery T-tube insertion in a patient with tracheal stenosis due to relapsing polychondritis (RP).

Case Report

A 24-year-old man diagnosed with RP came to our hospital with the chief complaint of dyspnea. During admission, pulmonary function tests showed severe airway obstruction (forced expiratory volume within the first second/forced vital capacity, 1.21 L/4.59 L=26%). Thoracic computed tomography (CT) demonstrated diffusely thickened and narrowed airway. The smallest diameter was 2.5 mm for the left main bronchus, 5 mm for the right main bronchus, and 5×10 mm for the trachea at the thoracic inlet. Bronchoscopy revealed a horseshoe-shaped trachea lumen extending from 1 cm below the vocal cord to 2 cm above the carina, and stenosis of bilateral main bronchi. A diagnosis of critical airway stenosis caused by RP was made. The stenosis of the left main bronchus, the narrowest part shown on CT, was thought to be the main reason for his respiratory symptom. Thus, endobronchial stenting of the left main bronchus was arranged and performed uneventfully. However, dyspnea developed again postoperatively. Due to persistent progressive dyspnea and the known tracheal stenosis, a second procedure, Montgomery T-tube insertion, was arranged.

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During the second operation, the surgeon first created a tracheostomy, and then a T-tube was inserted into the trachea. A 6.0 endotracheal tube was placed via the horizontal arm of the T-tube for ventilation. Severe airway obstruction was noted after the patient was ventilated via the 6.0 endotracheal tube. The airway was examined by flexible fiberoptic bronchoscopy, which revealed folding of the T-tube and a collapsed tracheal lumen. The trachea was dilated with dilators. However, airway obstruction was not resolved despite several attempts to reposition the Montgomery T-tube. The patient's oxygenation was maintained with pure oxygen initially, and airway pressure was kept at 28 mmHg under pressure control mode with variable tidal volume (320–580 mL). However, hypercapnia developed gradually, and end tidal carbon dioxide partial pressure gradually increased to > 90 mmHg. At this moment, hypoxia developed gradually, and pulse oximetry revealed a declining value to < 75%. The T-tube was pulled out and oral tracheal intubation was immediately performed. Nevertheless, the patient's condition deteriorated persistently with bradycardia and no detectable end-tidal carbon dioxide; cardiac arrest then occurred.

Cardiopulmonary resuscitation was performed immediately. Meanwhile, arterial blood gas analysis revealed severe hypoxemia and hypercapnia (pH 6.95, PaO_2 of 23 mmHg, $PaCO_2$ of 170 mmHg). Bilateral breath sounds were checked; they were barely heard and mingled with faint wheezing sounds, especially on the right side. Due to the persistent high airway pressure and refractory cardiac arrest, a cardiovascular surgeon was consulted for emergent extracorporeal membrane oxygenation (ECMO). During this time, tension pneumothorax was suspected. A chest tube was tried on the right, which released a large amount of air but did not lead to much improvement in the vital signs. Another chest tube was inserted on the left side, which also released a large amount of air and, more importantly, resulted in immediate improvement in the vital signs. After initiation of venovenous ECMO for gas exchange, the surgeon rechecked the tracheostomy site. A laceration over the membranous portion of the trachea 1–2 cm below the tracheostomy wound was found. Esophagoscopy was performed and revealed no esophageal injury. The surgeon repaired the laceration directly through the original tracheostomy. After several unsuccessful attempts at T-tube and extended-length tracheostomy tube insertion, the procedure for tracheal stenting was abandoned, and the tracheostomy was closed. At the end of the operation, it was noticed that the patient had a distended abdomen, but his vital signs were stable. The patient was ventilated via an oral endotracheal tube and sent to the intensive care unit under ECMO and intravenous dopamine support.

In the intensive care unit, the patient received positive pressure ventilation and ECMO support. Unfortunately, progressive tachycardia, hypotension and hypoxia developed again. In addition, the patient's abdomen was noted to be even more distended despite attempted decompression with both transanal and nasogastric tubes. Portable chest film revealed free intra-abdominal air and good expansion of bilateral lungs. Bedside abdominal sonography showed massive pneumoperitoneum; bowel perforation was tentatively ruled out because no fluid accumulation in Morrison's pouch and splenic fossa was found on sonographic examination. Conservative abdominal paracentesis with pigtail-drainage tube insertion resulted in immediate relief of abdominal distension and improvement in the patient's hemodynamics and oxygenation. A follow-up chest X-ray showed no free air in the abdomen.

ECMO was discontinued 2 days later. Unfortunately, follow-up brain CT confirmed hypoxic encephalopathy. Three months after his admission, the patient died of multiorgan failure secondary to sepsis.

Discussion

RP is a rare multisystem disorder of unknown etiology and is characterized by recurrent inflammation and destruction of cartilage and connective tissues.[3–6] It has been shown that airway involvement usually predicts a poor prognosis and is one of the leading causes of death in RP.[3–9] Airway involvement, manifesting mainly as airway obstruction,[3,5,7] as in our patient, also presents a significant challenge for airway management, especially when the airway inflammation is diffuse and cannot be controlled by medication. Fatal complications during airway management include complete airway obstruction and airway rupture.[6,10] However, there is no case report in the literature about these complications during anesthesia for airway procedures, which may be attributed to the rarity of this disease.[5,8] Nevertheless, the need for airway procedures and management will expose anesthesiologists to these rare and difficult patients.[11]

The development of tension pneumothorax and tension pneumoperitoneum in our case was presumably due to the tracheal tear. Multiple risk factors for airway laceration have been reported in the literature.[12,13] Factors such as operator errors (multiple attempts or inexperienced physicians), equipment selection (malposition of the tube, improper airway dilator

or tube size), and patient factors (steroid users, chronic obstructive/inflammatory pulmonary disease or tracheomalacia) may contribute to airway rupture. Most of these risk factors were present in our patient. Thus, a cautious approach should be emphasized during airway manipulation in patients with RP because they may be more vulnerable to airway trauma, especially when the inflammation of the airway is diffuse. Our patient underwent 2 consecutive airway operations in a short period of time. The stress and trauma induced by surgery and intubation might cause more inflammation and further obstruction in the airway. We think this may be the reason for why T-tube insertion failed and caused tracheal tear in our patient.

Tracheobronchial rupture with delayed diagnosis is a potentially fatal complication. It can lead to life-threatening tension pneumothorax, especially in anesthetized patients under positive pressure ventilation.[14] Early recognition is very important. However, diagnosis of such a condition may be difficult, especially when tension pneumothorax is bilateral. It is easily misdiagnosed as acute airway obstruction. Both acute airway obstruction and bilateral tension pneumothorax can present with increased airway pressure, oxygen desaturation, hemodynamic compromise and even cardiac arrest. In our case, the raised airway pressure and progressive hypercapnia, presented early before the cardiac arrest, was initially attributed to the collapsed airway distal to the Montgomery T-tube. The surgical drape, which hindered prompt physical examination including inspection for neck vein engorgement, the anesthetized patient, and the lack of a lateralizing sign in bilateral tension pneumothorax, all provided additional challenges to prompt diagnosis of bilateral tension pneumothorax. Thus, a high level of suspicion must be kept and bilateral tension pneumothorax should be considered during refractory cardiac arrest in all patients with increased airway pressure, particularly in patients with airway disease such as RP.

Pneumoperitoneum is usually a surgical emergency because it indicates bowel perforation in most cases. However, pneumoperitoneum can also result from thoracic causes, such as tracheal rupture and pneumothorax,[15–20] and can be managed conservatively[16] as in our patient. We believe that under positive ventilation and high airway pressure, the gases leaked from the ruptured trachea, down the passage created by the transhiatal dissection of the esophagus into the peritoneal cavity, and caused the pneumoperitoneum in our patient. Thus, tension pneumoperitoneum developing as a result of tracheal tear is a rare but life-threatening complication, but it can be easily managed if immediate diagnosis is made.

In conclusion, RP with airway involvement has serious implications for anesthesiologists and surgeons. The inflammatory airway may lead to both airway obstruction and airway rupture. Both anesthesiologists and surgeons must be prepared to deal with the possible complications that may occur during airway manipulation. These may include tracheal rupture complicated with bilateral tension pneumothorax and pneumoperitoneum. We emphasize that bilateral tension pneumothorax should be considered during refractory cardiac arrest in all patients with increased airway pressure. A high index of suspicion and adequate management are mandatory for patients to survive these life-threatening complications.

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